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AUTHOR(S):

Segawa, Naoki; Gohji, Kazuo; Masuda, Hiroshi; Katsuoka, Yoji

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A CASE OF CA19-9-PRODUCING SEMINAL VESICLE CYST WITH IPSILATERAL RENAL AGENESIS

Naoki SEGAWA, Kazuo GOHJI, Hiroshi MASUDA and Yoji KATSUOKA
From the Department of Urology, Osaka Medical College

A case of CA19-9-producing seminal vesicle cyst with ipsilateral renal agenesis is reported. A 29-year-old man was admitted to our hospital for perineal pain and urinary retention. Digital rectal examination revealed a large soft mass that fluctuated in the area of the prostate and seminal vesicles. Magnetic resonance imaging revealed a right kidney defect, and ipsilateral dilation and cystic enlargement of the right seminal vesicle. Transrectal puncture of the seminal vesicle cyst was performed. The contents were pus and old red blood cells. Initially, the serum CA19-9 level was extremely high (145.8 U/ml) but was normalized by the treatment with antibiotics after the puncture. The symptoms subsided without recurrence.

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Key words: Seminal vesicle cyst, Renal agenesis, Serum CA19-9

INTRODUCTION

The association of seminal vesicle cyst and ipsilateral renal agenesis was initially reported in 1914 by Zinner[1]. Unilateral renal agenesis occurs in 0.1% of newborns[2] Genital anomalies are found in 12% of men with unilateral renal agenesis. Congenital seminal vesicle cyst is associated with ipsilateral renal agenesis in 68%[3] Since then, approximately 50 reports have been published of this developmental abnormality[4–7] Its association with the production of CA19-9, however, is uncommon. We report a case of CA19-9-producing seminal vesicle cyst associated with ipsilateral renal agenesis.

CASE REPORT

A 29-year-old man was referred to our hospital on February 18, 2002 with chief complaints of perineal pain and urinary retention. Physical examinations, including some tests in the genital region, revealed a normal, healthy man. A digital rectal examination revealed a large, smooth (soft, fluctuant) mass arising from the area of the prostate and seminal vesicles. A few red blood cells were present on urinalysis. Blood-serum tests confirmed inflammatory change, such as an increase in the number of leukocytes (11,630/mm^3, normal range 3,300–8,190/mm^3) and CRP elevation (17.08 mg/dl; normal range$<$0.25 mg/dl). Laboratory evaluation revealed an elevated tumor marker CA19-9 (145.8 U/ml; normal range$<$ 37 U/ml). Magnetic resonance (MR) imaging of the abdomen and pelvis showed right renal agenesis and a large mass, measuring 8 cm in its greatest diameter, arising from the right seminal vesicle. The mass showed low T1-weighted and high T2-weighted signal intensity compatible with a cyst (Fig. 1). We found an anatomical structure that semed to be an

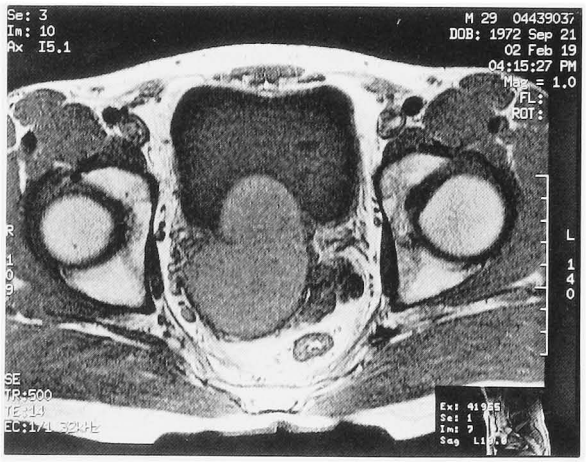

Fig. 1. MR imaging of the pelvis showed a large mass with high T1-weighted signal intensity, which measured 8 cm in its greatest diameter, arising from the right seminal vesicle.

ectopic ureter terminating into the mass. Cystourethroscopy revealed normal urethral mucosa and verumontanum with a distinct bulging of the right hemitrigone and absence of the right ureteral orifice. Therefore, right ectopic ureter opening into the seminal vesicle was suspected. He was initially treated with antibiotics and the inflammation was incompletely resolved. For diagnostic purposes, and as an alternative treatment, we performed puncture and aspiration of the cyst by the transrectal route and injected a contrast medium. The cystic mass was a dilated right seminal vesicle communicating with the right ureter (Fig. 2). The cystic tumor was well defined with no irregular thick wall. It contained approximately 100 ml brownish, viscous fluid, which contained many leukocytes, not fresh spermatozoa. The sample had a high concentration of CA19-9 (4,864 U/ml), but no malignant cells were found.

Fig. 2. Intraoperative contrast medium injection demonstrates the distal end of the right ureter dilated and communicating with the right seminal vesicle.

He was treated by instillation of an antibiotic into the cyst. He remained asymptomatic. Transrectal ultrasonography revealed the complete disappearance of the cystic lesion behind the bladder. The level of serum CA19-9 returned to the normal range 6 months after the puncture.

DISCUSSION

Congenital seminal vesicle cysts induced by obstruction of the ejaculatory duct are commonly associated with anomalies of the ipsilateral upper urinary tract or mesonephric duct, resulting in renal agenesis, hypoplasia, dysplasia, and ectopic ureteral opening to the seminal vesicle or a defective trigone. They usually involve pain on intercourse, bladder irritation or epididymitis in the years of maximal sexual activity[3].

The treatment choice for a seminal vesicle cyst depends on the symptoms related to size and location. Surgical intervention is justified in patients who fail to respond to more conservative treatment options (antibiotics, aspiration of the cyst, transurethral deroofing of the cyst). If malignancy cannot be excluded, histological examination of the cyst is necessary. Aspiration of the cyst should be performed initially because 30% of patients are cured, though it is considered only as a diagnostic procedure due to the high failure rate and the risk of infection. In our case it was successful. However, a longer term follow-up is necessary.

The CA19-9 antigen has become the most useful blood test in the diagnosis and management of patients with cancer of pancreatic and gastrointestinal carcinoma[8–11]. It is a tumor-associated, not a tumor-specific, antigen. It is well known that CA19-9 expression is identified in 76% of transitional cell carcinoma[12] and the serum level is elevated in some nonmalignant diseases. Moreover, it has been found in normal seminal fluid[13]. The expression in the normal renal pelvis and ureter has been reported in Japan[14]. Some rare hydronephrosis and hydroureter cases with extremely high serum levels have also been described[15–17] The aspirated fluid contained a high level of CA19-9. Our case implies that a seminal vesicle cyst and hydroureter may cause elevated serum CA19-9. A case of seminal vesicle cyst producing CA19-9 with ipsilateral renal agenesis is rare. To our knowledge, this combination has been described in only one previous report[18] It is thought that the production of CA19-9 from the epithelium of the seminal vesicle is stimulated by inflammation[19], although the pathway or process by which CA19-9 traverses between seminal fluid and the blood compartments is unknown. One possible mechanism is that CA19-9 in the seminal fluid is transferred to the serum due to damage of vessel wall by inflammation. In our case, the serum level dramatically decreased after aspiration of the cyst and became normal. Thus, the level of CA19-9 might serve as a good index for observing the clinical course of the patient.

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和文抄録

同側腎無形成を合併したCA19-9産生精嚢嚢胞の1例

大阪医科大学泌尿器科学教室（主任：勝岡洋治教授）
瀬川　直樹，郷司　和男，増田　　裕，勝岡　洋治

今回，われわれは同側腎無形成を合併したCA19-9産生精嚢嚢胞の1例を報告する．症例は29歳，男性，残尿感，会陰部痛を主訴として受診した．直腸診にて波動を有する前立腺を触知した．MRIでは右腎は欠損し，同側の精嚢は拡張し嚢胞状変化を伴っていた．経直腸的精嚢穿刺術を施行し，内容液は血性膿汁であった．初診時，血清CA19-9が145.8 U/mlと異常高値であったが抗生剤投与と穿刺術にて正常化し，症状も消失している．

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